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Family Patterns of Developmental Dyslexia Part III: Spelling Errors as Behavioral Phenotype

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The major trends in current research on developmental dyslexia assume that impaired phonological processing is the core deficit in this disorder. Our earlier studies indicated that half of all dyslexic persons have significant deficits of bimanual motor coordination, and that impaired temporal resolution in motor action may identify a vertically transmitted behavioral phenotype in familial dyslexia. This report examines the relationship between spelling errors as a measure of impaired phonological processing and motor coordination deficits in the same dyslexia families. Affected family members *with* motor coordination deficits made significantly more dysphonetic spelling errors than dyslexic family members *without* motor deficits, but there was no evidence that dysphonetic spelling is vertically transmitted in dyslexia families. On the other hand, affected offspring of affected parents with *motor coordination* deficits made relatively more dysphonetic spelling errors than the affected offspring of parents without motor coordination deficits. We suggest that dysphonetic spelling may be one outward expression of a vertically transmitted behavioral phenotype of impaired temporal resolution that is clearly expressed in coordinated motor action. © 1996 Wiley-Liss, Inc.

KEY WORDS: familial dyslexia, dysphonetic spelling, impaired temporal resolution

INTRODUCTION

Developmental dyslexia is an etiologically and functionally heterogeneous condition [Ellis, 1985], and some subtypes are thought to have a genetic etiology. However, the exact mechanisms of inheritance remain an issue of enduring controversy [Decker and Bender, 1988; Finucci et al., 1976; Hallgren, 1950], and the behavioral phenotypes that are vertically transmitted in familial dyslexia remain essentially unknown [Cardon et al., 1994; Rabin et al., 1993; Smith et al., 1983].

The presenting symptom of reading impairment in dyslexia is usually part of a multifaceted syndrome that also includes linguistic, cognitive, perceptual, and motor deficits in various combinations [Denckla, 1993]. Some associated behavioral deficits are probably coincidental findings, but others may identify domain-general functional deficits as well as behavioral phenotypes that are transmitted in familial subtypes.

An extensive body of experimental and clinical evidence supports the hypothesis that impaired phonological awareness is *the* core deficit of developmental dyslexia [Ellis, 1985; Fletcher et al., 1994; Share, 1995; Vellutino, 1979; Wagner and Torgensen, 1987]. However, most of the supporting evidence has been collected in English-speaking students [Caravolas, 1993]. Since English orthography is generally recognized to be one of the "deepest" and most difficult to master for beginning readers [Ehri, 1993; Katamba, 1994], impaired phonological processing may be the core deficit of dyslexia among English speakers, whereas other language deficits may be the primary source of reading impairment in languages with transparent orthographies [Bugarski, 1993; Caravolas, 1993; Ehri, 1993].

Although poor readers in such languages also show some degree of impaired phonological awareness when first learning to read, they usually compensate for this deficit within the first 2 years of schooling, but continue to be poor readers [Cossu et al., 1988; Lindgren et al., 1985; Valle-Arroyo, 1987]. For example, the phoneme-grapheme correspondence rules of German are more regular and transparent than those of English, and more complex than those of Spanish or Italian. German-speaking children who are poor readers at time of school entry do have difficulty decoding phonemically-mediated words, but usually overcome this deficit within 2 years of schooling. Nevertheless, they continue to be poor readers, the primary source of their difficulties being substantially slower reading rates and longer latencies on lexical retrieval tasks [Wimmer, 1993; Wimmer et al., 1991]. Similarly, reading-impaired Spanish-speaking students quickly

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compensate for their early phonological processing deficits, but continue to read at significantly slower rates [Valle-Arroyo, 1987]. A reduced rate of processing language may therefore be the primary source of difficulty for dyslexic students in languages with transparent orthographies. In fact, even among English-speaking impaired readers, prolonged naming latencies and reduced reading rates may identify a distinct, although overlapping, subtype of dyslexia [Bowers and Wolf, 1995].

The point of departure for our dyslexia studies was the assumption that language deficits may be a major proximal cause but that they are not self-explanatory, and that they imply "underlying" functional deficits that transcend national variations of phoneme-grapheme correspondence rules. More specifically, we investigated impaired temporal resolution in motor action as such an underlying functional impairment. On one hand, variables of frequency, timing precision, and serial ordering are fundamental organizing principles of central nervous system function [Llinas, 1993]. On the other hand, "slow information processing," "sequencing difficulties," and impaired language prosody are frequently cited as major nonreading correlates of the reading retardation in dyslexia [Deckla, 1979].

Although clinically-observed sequencing difficulties have rarely been defined operationally, experimental support for such a hypothesis comes from studies by Lovegrove et al. [1986] and Lovegrove [1993], and by Tallal et al. [1985a,b], who examined temporal processing of nonlinguistic visual and auditory stimuli arrays. Their findings converge on the conclusion that the speed and efficiency of temporal information processing of perceptual events discriminate efficiently between dyslexic or language-impaired students and normally achieving students.

We focused on timing in coordinated motor action because experimental evidence from many sources converges on the conclusion that the neural control of temporal organization in speech and language overlaps extensively with dimensions of response frequency, timing precision, and serial ordering in fine motor skills [Keele et al., 1990; Kelso and Tuller, 1981; Kimura and Archibald, 1974; MacNeilage, 1987; Ojemann, 1984]. At the same time, coordinated motor action is uniquely well-suited for the experimental analysis of temporal dimensions in behavioral organization, without totally degrading the phenomena of primary interest [Lashley, 1951; Sperry, 1952].

In cross-sectional studies, we showed that the timing precision in half of all dyslexic students from age 8–25 years is significantly impaired on tasks of bimanual motor coordination and motor speech repetition, relative to age-matched normal readers [Wolff et al., 1990a,b, 1995; Wolff, 1993]. Subsequent studies of dyslexia families have indicated that half of reading-impaired relatives showed significant motor coordination deficits, whether or not they had been diagnosed as being dyslexic. Moreover, affected relatives with motor deficits have come from dyslexia families where probands also have significant motor deficits, while affected relatives without motor coordination deficits have come from families in which probands do not have motor deficits. Finally, affected offspring with two affected parents were twice as likely to show impaired timing control in bimanual and motor speech tasks as affected offspring with one affected parent [Wolff et al., 1995].

On the basis of these and related findings, we concluded that impaired temporal resolution in motor action may identify a developmentally invariant behavioral phenotype in familial dyslexia that is transmitted as a dominant trait. Because of the widely-accepted hypothesis that impaired phonological processing identifies a core deficit in developmental (and presumably familial) dyslexia, we tested the relationship between phonological processing deficits and impaired temporal resolution of motor action in the same group of dyslexia families.

MATERIALS AND METHODS

Sample

The demographic and behavioral characteristics of families selected for study have been described in detail in earlier reports [Wolff and Melngailis, 1994; Wolff et al., 1995]. Families were ascertained through probands attending specialized schools and a college for dyslexic students that only accepts students after an intensive review of previous school records, standardized intelligence tests, reading, writing, and spelling skills, focused language tests, and a physician's report [Wolff and Melngailis, 1994; Wolff et al., 1995]. Minimum admission criteria to these schools include average or above average psychometric intelligence, no uncorrected sensory deficits, and no major organic, neurological, or primary emotional difficulties, but significant deficits in reading and writing despite exposure to an adequate learning environment.

Families were asked to participate in the empirical studies only if probands scored at or above 100 on standardized intelligence tests, and if, depending on their age, probands were reading *and* spelling at least 2 or 3 years below grade level. Although lower IQ scores or less severe reading and spelling impairment are obviously compatible with a diagnosis of developmental dyslexia, the stringent selection criteria were adopted to increase the likelihood that probands in these families were "truly dyslexic."

All families meeting the criteria who agreed to participate received questionnaires that elicited information about a) the family's demographic characteristics, b) the age, sex, hand preference, highest academic grade reached, and history of difficulties in learning to read, of each first- and second-degree relative, and c) each relative's *current* reading, spelling, or other academic difficulties, reading habits, occupation, and whether any of them had ever repeated a grade. Respondents were encouraged to discuss their answers with relatives and to consult available records before answering questions. They were also asked to indicate how certain they were about the responses, and to elaborate on their responses with narrative answers whenever they could. Arithmetic difficulties were recorded but not used as a basis for classifying family members. Socioeconomic status was scored by the Hollingshead-Redlich four-factor scale [Hollingshead, unpublished manuscript, 1975].

On the basis of the questionnaire data, all first- and second-degree relatives were classified as affected by history if they reported significant difficulties learning to read during their school years [see Pennington et al., 1986; Wolff and Melngailis, 1994, for rationale]; and each family was assigned to 1 of 4 pedigree types by criteria summarized in Table I. All families fitting any one of the four pedigree types were asked to participate in the experimental study.

Appropriateness of the pedigree classification was checked in greater detail by interviewing all first-degree relatives in participating families in greater detail about the nature and severity of their reading, spelling, and other academic difficulties, reading habits, and other potentially informative personal data. Families who failed to meet pedigree criteria on the basis of these interviews were reassigned to a different category if appropriate, or dropped from the study; additional families were recruited until at least 20 families of each pedigree type had agreed to participate. All adults in the sample had completed high school, and no special provisions were made to correct for school dropouts [Finucci et al., 1976].

Participating subjects were examined by a detailed experimental protocol as well as by academic achievement tests. First-degree relatives of normal intelligence (WISC-R or WAIS-R scores >90) who were below age 12 years were classified as affected by *testing* if they were reading *and* spelling at least two grades below grade level; those between age 12–18 years, if they were reading *and* spelling at least three grades below grade level; and those above age 18 years, if they were *either* reading *or* spelling at least three grades below a twelfth grade ceiling. The reasons for choosing different criteria to classify persons below and above age 18 have been detailed in Pennington et al. [1986] and Wolff et al. [1995].

The final sample was initially divided into six major age groups that corresponded closely to the actual age distribution of probands and relatives. For this report, subjects were reclassified into four age groups of equal size: children below age 13, adolescents from age 13–18, young adults from age 18–40, and adults over age 40.

TABLE I. Families Defined by History and Testing*

	Type of family			
	B	P	M	PO
Families (N)	25	22	26	20
Parents (N)	50	44	52	40
Sisters (N)	15	22	19	21
Brothers (N)	22	15	18	15

* B families, proband and both parents affected; relatives on both sides may be affected. P families, proband and father affected; relatives on father's side of family may be affected; neither the mother nor any of her relatives are affected. M families, proband and mother affected; relatives on mother's side of family may be affected; neither the father nor any of his relatives are affected. PO families, proband affected; neither parent affected, no siblings affected; no aunts, uncles, or grandparents affected.

Protocol

Classifying measures. Probands were not retested for psychometric intelligence or academic achievement, because all of them had been examined in detail at their schools during the preceding 12 months. First-degree relatives were tested on a short version of the WISC-R or WAIS-R that correlates reliably with the full Wechsler scale [Sattler, 1974], and by the *Gray Oral Reading Test* and the *WRAT Spelling Subtest* (level I or II, depending on age).

Experimental measures. *Spelling errors.* Level I or II of the *WRAT Spelling Subtest* was administered to all participating family members, and they were encouraged to spell as many words as possible, even when they did not know the spelling or the meaning of the word. Spelling errors were scored globally as *phonologically acceptable* or *dysphonetic* by criteria similar to those used by Finucci et al. [1983]. A misspelled word was scored as *phonologically acceptable* if it could be pronounced to sound like the target word by analogy or according to loosely-applied phonics rules. Misspelled words were scored as *dysphonetic* if a) they failed to meet the above criterion; b) a syllable was either omitted, or added, or the sequence of syllables in a word was reversed; c) a wrong syllable was substituted or inserted; or d) subjects spelled an entirely different word, whether or not they spelled it correctly. By these criteria, the misspelling of "heaven" as *hieven* (as in "friend"), of "nature" as *nacher,* of "exaggerate" as *exagerate,* or of "majority" as *mugarity* were scored as phonologically acceptable. The misspelling of "lucidity" as *lucity,* of "majority" as *mugaratory,* or of "effeminate" as *effinemate* were scored as dysphonetic.

Before analyzing test records, two judges scored the protocols of 10 dyslexic students who were not part of the study by these criteria. After reconciling their initial disagreements, they reached a >95% interscorer reliability. All errors were also scored qualitatively according to categories that were created for this study. Although many schemes have been proposed for analyzing spelling errors qualitatively, each scheme differs to a greater or lesser degree from the others [Holmes and Peper, 1977; Moats, 1983; Roeltgen, 1992]. Rather than attempting to reconcile these differences, we therefore tabulated all spelling errors, grouped them by error type, and derived a qualitative taxonomy of spelling errors, summarized in Table II, that allowed us

TABLE II. Taxonomy of Spelling Errors

Phonologically acceptable spellings
1. Wrong vowels
2. Wrong consonants
3. Wrong vowels and consonants

Dysphonetic spellings
1. Syllable deletions: first, middle, or last
2. Syllable additions: first, middle, or last
3. Letter/syllable reversals
4. Wrong syllables
5. Wrong word but orthographically correct

to classify all errors reliably, except for a few with bizarre spellings that could not be classified.

Interlimb coordination. Methods of data collection and data reduction, and analysis of bimanual coordination tasks, have been described in detail previously [Wolff et al., 1995], and are summarized here. Subjects were instructed to tap two 5 × 3 cm touch-sensitive plates mounted on a Bakelite surface by moving the index finger of each hand at the metacarpal-phalangeal joint while resting the other fingers, the hands, and the wrists on a supporting cushion on the table surface.

They tapped their index fingers in time to a computer-generated metronome signal that continued for the first 10 sec of each trial and then stopped, while they continued tapping at the same rate and in the same pattern for another 20 sec. Only the performance of the last 20 sec (continuation condition) was entered for statistical analysis. Practice trials at a slow rate were given until subjects could perform the task as prescribed.

The protocol included 1) tasks of *symmetric alternation* at four different response frequencies, for which subjects had to alternate responses by the two index fingers in a stable alternating rhythm; 2) *asymmetric* tasks of bimanual coordination at four different response frequencies, for which subjects had to tap with the right "leading" finger in time to every beat of the metronome, while the "nonleading" left finger responded to every *other* metronome signal, so that the two fingers tapped in unison on every other beat; and 3) *asymmetric* tasks of bimanual coordination at four different response frequencies, for which subjects had to tap with the left leading finger in time to every beat of the metronome while the nonleading right finger responded to every other beat. Timing precision was measured as the standard deviation of interresponse intervals (IRI), and was modified to correct for departures from "stationarity" (e.g., when subjects gradually slowed down or speeded up during the course of a trial); the modified statistic is here identified as the acceleration standard deviation (ASD).

Our earlier cross-sectional studies had shown that relatively simple bimanual tasks performed at the slower response rates discriminated efficiently between *young* dyslexic students and age-matched normal controls, but were no longer discriminatory at older ages. The same tasks at a faster response rate, or more difficult bimanual tasks, discriminated efficiently between older dyslexic subjects and age-matched normal controls, but were not discriminatory at younger ages because they were too difficult for nearly all subjects. The dimension of behavioral coordination that best captured the functional differences between dyslexic persons and normal readers was the response frequency at which subjects could maintain a prescribed motor pattern with relative stability of timing precision. As the findings indicated, the particular frequency threshold for timing precision varied with age, but the general phenomenon of impaired temporal resolution in motor action was developmentally stable. Because the dyslexia families for these studies included individuals ranging in age from 7–60 years, the protocol of bimanual tasks included 12 individual tasks at different levels of difficulty (i.e., different response frequencies, different patterns of bimanual coordination, or both), to insure that at least one measure of the protocol would be in the appropriate range of difficulty for each age group.

Statistical analysis of the findings in this extensive protocol presented methodological problems, because many of the bimanual tasks were highly intercorrelated. To avoid computational confounding due to collinearities, we derived ordinal measures of motor coordination that would allow us to compare reading groups across age groups. For each age group, the single task of bimanual coordination was identified that discriminated most efficiently between affected family members and biologically-unrelated, age-matched normal controls. On the basis of these age-specific tasks, a score of 1 was assigned to each affected or nonaffected family member who performed at <1 SD above mean performance of age-matched normal controls; a score of 2 to each family member who performed between 1–2 SD above the mean performance of normal controls; and a score of 3 to those who performed at 2 SD or more above normal controls.

A frequency tabulation indicated that nearly all subjects received scores of either 1 or 3, while <7% of all family members received scores of 2. Scores of 2 and 3 were therefore collapsed into a single binary score of + that identified subjects with significant motor deficits, while a score of − identified subjects with no significant motor deficits.

Motor speech. Subjects were instructed to repeat 10 three-syllable strings (pa-ta-ka) and 15 two-syllable strings (ta-ka) at various rates specified by a metronome signal, and to pace their speech repetitions so that the first syllable in either the two-syllable string (ta) or the three-syllable string (pa) coincided with each metronome signal. Speech samples were recorded on audio tape and analyzed off-line for 1) total time to complete the requisite number of sequences; 2) deviations in either direction from the time needed to perform the prescribed tasks at the expected rate; and 3) speech sequencing errors (syllable duplications, omissions, order reversals, and intrusion of extraneous syllables [see Wolff et al., 1990b]). Practice trials were given at a slow rate until subjects understood and could perform the task at the slow rate.

For reasons outlined above, the full motor speech repetition protocol also included six separate tasks at different response frequencies or levels of difficulty. Because some individual tasks were again highly intercorrelated, a similar strategy was adopted to classify each family member by a motor speech score of + or −.

RESULTS

Spelling Errors

By definition, affected relatives and probands made more spelling errors, and spelled fewer words altogether, than nonaffected family members. The total number of spelling errors was therefore computed as a *percentage* of all words attempted. Similarly, the frequency of *dysphonetic* errors was computed as percent-

age of all spelling errors. One-way Analyses of Variance (ANOVAs) (SAS/STAT, GLM model, 1990), with affection status as classifying variable, and with mean number of words spelled, percentage of all errors, and percentage of *dysphonetic* errors as dependent measures, indicated that dyslexic family members spelled fewer words but made a larger percentage of spelling errors. Affected persons also made more dysphonetic errors as percentage of all spelling errors (see Table III).

Age and sex effects. Similar ANOVAs in affected persons, with sex and age group as classifying variables, indicated that there were main effects by sex for total number of words spelled (F (1, 285) = 14.9, P = .0001) and for percentage of errors (F (1, 284) = 7.1, P = .008), but not for percentage of dysphonetic errors. Affected females spelled more words altogether than males, while affected males made proportionately more spelling errors. There were also main effects by age for percentage of dysphonetic errors (F (3, 281) = 8.3, P = .0001), but not for total words or percentage of all errors; and age effects were significant only between children under age 13 years and all others ($P < .05$, by Duncan post hoc multiple comparisons). There were no sex by age group interactions.

Parallel ANOVAs on nonaffected family members indicated that there were no main or interaction effects by sex. Age did, however, have a main effect for percentage of errors (F (3, 204) = 3.9, P = .0023), and for percentage of dysphonetic errors (F (3, 203) = 6.44, P = .0001). Normally reading children spelled as many words as adults, but they made expectedly more spelling errors and a larger percentage of dysphonetic errors ($P < .05$ by Duncan multiple comparisons). This result was expected, because nearly all subjects were tested by level II of the *WRAT Spelling Subtest* that includes many words intended to be too difficult for children, and all subjects were encouraged to spell all words. However, there was no a priori reason for assuming that children would also make relatively more dysphonetic errors than adults.

Reading delay and spelling deficits. Reading and spelling deficits are highly correlated among dyslexic students of elementary school age, but many older dyslexic persons compensate for their earlier reading difficulties while continuing to be poor spellers, so that reading and spelling scores may become increasingly dissociated with age [Frith, 1986; Pennington et al., 1986; Wolff and Melngailis, 1994]. To test whether this dissociation was nonspecific or applied specifically to *dysphonetic* spelling errors, we divided all affected persons within each age group into subgroups who scored above or below the mean reading level of affected persons of the sample. ANOVAs with age group and relative severity of reading impairment as classifying variables, and with percentage of dysphonetic errors and of total errors as dependent measures, indicated that there were main effects by reading impairment (F (1, 287) = 29.6, P = .0001) and age group (F 93, 287) = 14.34, P = .0001) for percentage of dysphonetic errors, but reading group by age group interactions. Parallel ANOVAs for percentage of all spelling errors indicated that in addition to the expected main effects by reading group (F (1, 287) = 114.6, P = .0001) and age group (F (3, 287) = 46.4, P = .0001), there were also significant reading group by age group interactions (F (3, 287) = 13.4, P = .0001). In other words, the disposition to make dysphonetic errors was developmentally stable, even in persons who compensated for earlier reading disabilities, while overall spelling performance improved more or less in tandem with reading performance among dyslexic adults who had compensated for earlier reading difficulties.

Subtypes of spelling errors. A qualitative analysis of spelling errors according to the categories listed in Table II indicated that the *relative* distribution of dysphonetic error types did not differ between affected and nonaffected family members. Reading groups differed only with respect to the absolute numbers of dysphonetic errors and their subtypes, with respect to the absolute number of all spelling errors, and with respect to the distribution of errors on phonologically acceptable misspellings (see Table IV, and Fig. 1). On the latter, affected family members made twice as many vowel as consonant errors, while nonaffected relatives made equal numbers of vowel and consonant errors (χ^2 = 14.3, 1 df, P = .0001) [see also Bryson and Werker, 1989; Share, 1995].

Vertical Transmission of Spelling Errors

One major issue addressed in this report was the vertical transmission of dysphonetic spelling in familial dyslexia. As a first step toward addressing this issue, all affected parents and affected offspring were classified into subgroups making 0–20%, 21–40%, 41–60%, 61–80%, or >80% dysphonetic spelling errors. Thereafter, contingency tables were computed comparing the distribution of dysphonetic errors in affected parents and affected offspring. The results indicated that the affected offspring of affected parents with many dysphonetic spelling errors also made relatively many dysphonetic errors (χ^2 = 27.6, 16 df, $P < .05$).

However, the effect was modest, and group differences may have been inflated by converting continuous measures of dysphonetic spelling into ordinal scores.

TABLE III. Types of Spelling Errors in Affected and Nonaffected Family Members

	Affected (N = 287), mean	Not affected (N = 204), mean	F (1, 489)	P
Total words spelled	42.4	45.6	32.2	.0001
Errors/words spelled	0.573	0.264	377.1	.0001
Dysphonetic errors	10.8	2.4	190.2	.0001
Dysphonetic/all errors	0.41	0.16	174.3	.0001

TABLE IV. Subtypes of Spelling Errors

	Affected (N = 287)	Not affected (N = 204)
Phonologically Acceptable Words Errors		
Consonant errors	21%	36%
Vowel errors	36%	38%
Consonant + vowel errors	43%	25%
Dysphonetic Words		
Deletions: first syllable	0.3%	0.7%
Deletions: middle syllable	31.6%	27.5%
Deletions: last syllable	2.7%	1.3%
Additions: first syllable	0	0.7%
Additions: middle syllable	6.4%	11.8%
Additions: last syllable	0.9%	0.2%
Letter sequence reversals	8.6%	10.6%
Wrong syllables	45.09%	37.4%
Wrong words	4.6%	9.9%

Simple regression analyses were therefore computed with percentage of dysphonetic spelling errors by affected parents as the predictor, and percentage of dysphonetic spelling errors by affected offspring as the dependent variable. The dysphonetic spelling errors of parents accounted for only a small proportion of the variance in dysphonetic spelling errors of affected offspring ($R^2 = 0.026, P = 0.031$). Moreover, the pattern of association in affected family members was no greater than that found in nonaffected family members ($R^2 = 0.031, P = 0.0434$).

Pedigree effect. Dyslexia pedigree effects (see Table I) on dysphonetic spelling errors in affected offspring were tested by an ANOVA, with pedigree (both or only one parent affected) as classifying variable, and with percentage of dysphonetic spelling errors and percentage of all spelling errors in offspring as the dependent measures. There were main effects by pedigree for percentage of dysphonetic errors ($F(1, 129) = 9.45, P = .003$), but not for percentage of total spelling errors. Affected offspring with two affected parents made a larger percentage of dysphonetic errors (50%) than affected offspring with one affected parent (39%). Post hoc comparisons indicated that the only significant pedigree effects were between offspring with two affected parents and those with an affected mother (M families).

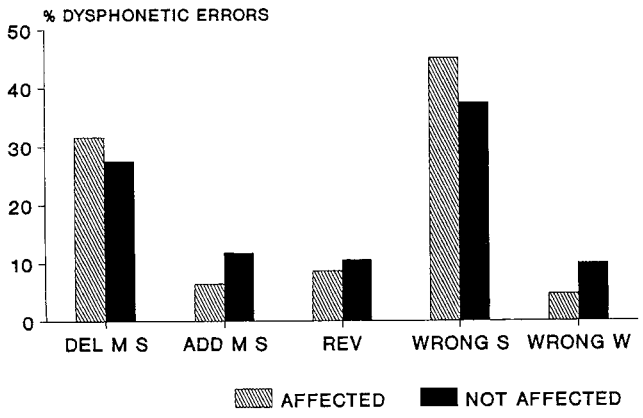

Fig. 1. Dysphonetic error types as percentage of dysphonetic errors. Del MS, delete middle syllable; Add MS, add middle syllable; Rev, reversals; Wrong S, wrong syllable; Wrong W, wrong word.

On the other hand, our previous studies indicated that dyslexic offspring with two affected parents are more severely impaired on measures of reading, spelling, motor coordination, and automatized naming than those with one affected parent; and that the *parents* in B families (both parents affected) are also more severely impaired in reading, spelling, and motor coordination than affected mother or fathers in M and P families, respectively [Wolff et al., 1995]. The results of this report indicated that parents in B families also made more dysphonetic errors than affected parents in M or P families ($F(1, 94) = 4.8, P = .031$). Thus, the apparent pedigree-specific effects on dysphonetic errors in offspring may have been confounded by the overall severity of impairment in both parents and offspring.

Relationship of Motor Deficits and Spelling Errors

The second main issue addressed in this report was whether motor coordination deficits cosegregate with dysphonetic spelling, or whether the two behavioral phenotypes identify distinct subtypes of familial dyslexia.

Within-subject relations. For reasons summarized earlier, binary scores of + or − were assigned to all family members to indicate whether or not they had significant deficits of bimanual and motor speech coordination.

As a first step toward testing the relationship between motor deficits and dysphonetic spelling, we computed ANOVAs in affected family members, with the two sets of binary motor scores as classifying variables, and with the percentage of dysphonetic and total spelling errors as dependent measures. Table V shows that there were main effects by both bimanual and motor speech scores for percentage of dysphonetic errors and percentage of all spelling errors. Family members *with* bimanual motor coordination deficits made relatively more dysphonetic errors and spelling errors of all kinds than affected members *without* motor coordination deficits. Similarly, affected family members *with* motor speech timing deficits made relatively more dysphonetic spelling errors than those without motor

TABLE V. Effect of Motor Scores on Spelling Errors of Affected Family Members (N = 286)

	Score			
Bimanual coordination	−	+	*F*	*P*
% errors/all words	40%	53%	10.5	.0013
% dysphonetic/all errors	26%	40%	19.3	.0001
	Score			
Motor speech	−	+	*F*	*P*
% errors/all words	55%	59%	NS	
% dysphonetic/all errors	35%	45%	13.5	.003

speech deficits; but there were no subgroup differences in overall spelling errors. Parallel analyses on nonaffected family members indicated that neither bimanual nor motor speech scores were correlated with either type of spelling error.

To control for the possible inflation of group differences by converting continuous motor measures to binary scores, we computed stepwise multiple regressions, using either the four most informative bimanual coordination tasks or the two most informative speech repetition tasks, as predictor variables, and the percentage of dysphonetic errors as the dependent measure. For reasons summarized earlier, it was to be expected that the proportion of variance explained by motor tasks would differ in the various age groups. Regressions were therefore computed both across and within age groups.

The analyses across age groups, summarized in Table VI, indicate that bimanual motor coordination and motor speech repetition tasks each accounted for a significant proportion of variance in dysphonetic spelling in affected family members. Table VII further shows that both sets of motor coordination variables also accounted for a significant proportion of variance in dysphonetic spelling in most age groups. As expected, the proportion of variance explained and the motor tasks that entered the regression equations differed across age groups. The R^2 in Tables VI–VIII is reported only for the first motor variable to enter the equation, which in fact accounted for >90% of the systematic variance in all cases.

As Table VI shows, motor coordination tasks also accounted for a small but statistically significant proportion of variance in dysphonetic spelling among nonaffected relatives, but none of the predictors contributed to the variance when regressions were computed separately in the four age groups. Earlier analyses indicated that dysphonetic errors per se are probably not vertically transmitted in dyslexia families, but our guiding hypothesis predicted that the *motor deficits* of affected parents would correlate with *dysphonetic spelling* in affected offspring. Additional stepwise regressions were therefore computed, with informative tasks of bimanual or motor speech coordination of affected parents as predictors, and with dysphonetic spelling errors of affected *offspring* as the dependent measure. Table VIII shows that parental bimanual coordination and motor speech variables predicted a significant proportion of variance in dysphonetic errors of offspring in affected members of dyslexia families. No such association was found in nonaffected family members.

TABLE VI. Relationship of Motor Variables and Dysphonetic Spelling (D%) Across Age Groups

	R^2	*F*	*P*
Affected (N = 258)			
Bimanual	0.138	41.0	.0001
Motor speech	0.129	34.4	.0001
Nonaffected (N = 190)			
Bimanual	0.018	5.15	.024
Motor speech	0.024	6.18	.016

TABLE VII. Relationship of Motor Variables and Dysphonetic Spelling by Age Group (D%)

		Affected subjects		
Bimanual Age group	N	R^2	*F*	*P*
1	76	0.161	11.4	.0008
2	73	0.198	18.1	.0001
3	66	0.049	2.65	.106
4	60	0.136	10.1	.002
Motor speech				
1	78	0.240	24.2	.0001
2	73	0.163	14.0	.0004
3	66	0.096	6.4	.009
4	65	0.051	3.5	.068

DISCUSSION

The main findings of this report are as follows:

1. Impaired temporal resolution in bimanual coordination and motor speech tasks is significantly correlated with overall frequency of spelling errors, and more importantly with percentage of dysphonetic errors in affected members of dyslexia families. The relationship was significant when tested either by categorical motor scores or continuous motor variables.
2. There was no evidence for a vertical transmission of dysphonetic spelling errors in dyslexia families.
3. However, the motor coordination deficits of affected parents were correlated with percentage of dysphonetic spelling errors in affected offspring. No such association was found in nonaffected parents and their nonaffected offspring.
4. Affected offspring with two affected parents made more dysphonetic errors than affected offspring with one affected parent, but the apparent pedigree effects could not be dissociated from relative severity of affection as the transmitted variable.
5. The qualitative spelling errors made by dyslexic and nondyslexic family members were the same in nearly all cases; most of the differences were only quantitative [see also Bryson and Werker, 1989; Holmes and Peper, 1977; Stage and Wagner, 1992; Treiman et al., 1993].

TABLE VIII. Parental Motor Deficits and Dysphonetic Spelling in Offspring

	Affected offspring		
Group	R^2	*F*	*P*
Affected parents (N = 118)			
Bimanual coordination	0.156	12.6	0.0004
Motor speech	0.072	6.4	0.012
	Nonaffected offspring		
Nonaffected parents (N = 67)			
Bimanual coordination	No variable entered in the equation		
Motor speech	No variable entered in the equation		

The combined findings of this and our earlier reports on the same families converge on the conclusion that impaired temporal resolution in motor action identifies one vertically-transmitted behavioral phenotype in familial dyslexia, and that this trait is functionally associated with a disposition to make dysphonetic errors. However, all of the inferences regarding impaired phonological processing were based on dysphonetic spelling errors produced while subjects spelled standard word lists to dictation. By contrast, most of the evidence on impaired phonological processing has come from experimental measures of phonemic segmentation, experimentally selected real words and nonwords, and the like. Focused investigations using such experimental measures are therefore needed before it can be concluded that motor measures of temporal resolution and dysphonetic spelling identify the same behavioral phenotype in familial dyslexia. To the extent that dysphonetic spelling identifies phonological processing deficits, the findings do, however, suggest that a common physiologically plausible dysfunction induces spelling, reading, and motor coordination deficits in familial dyslexia.

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